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Case Report : Two Cases of Juvenile Polyp

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Introduction

Juvenile polyp is a common disease and a major cause of rectal bleeding, next to anal fissure and intussusception in infants in other countries, but in Japan, only 34 cases of juvenile polyp had been reported by 1975⁴⁾. Recently the authors treated two patients with juvenile polyps.

Case presentation

Case 1 : The patient was a 6-year-old boy who for about one year had sometimes noted "ketchup-like" rectal bleeding during defecation, but had no abdominal pain, diarrhea, vomiting or any other complaints. A diagnosis of enteritis was made at another clinic where he was treated for three months.

He was introduced to another hospital for endoscopic and barium enema examinations, because of no improvement with medication and the suspicion of polyp in the colon. An exact diagnosis of polyp could not be made and he was discharged. The patient was seen at this hospital in January, 1978. There was no anemia or any other abnormality in laboratory tests. Barium enema showed a possible tumor shadow in the sigmoid colon, 2×2cm in size, as shown in Figure 1, and endoscopic examination was performed twice. In the sigmoid colon about 30cm from the anus, a solitary reddish smooth-surfaced pedunculated polyp was found on the second trial, and we decided to operate.

Case 2 : The patient was a 3-year-old boy who had rectal bleeding for three months, but no other symptoms. He visited this hospital in September 1978, and polyp



Fig. 1. X-ray finding of solitary polyp in sigmoid colon. (Case 1)

Key words : Rectal bleeding, Polyp of colon in children, Adenomatous polyp, Juvenile polyp

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was suspected. The barium enema examination showed a solitary movable polyp in the sigmoid colon. Since chronic blood loss and the resultant general debility would certainly be a hazard to the health of a growing child, we decided on transabdominal polypectomy. Endoscopic examination before operation was not performed, because of the difficulty and the risk of this examination in small children. Laboratory tests showed no anemia or any other abnormalities.

Operative findings

Both operations were performed through a low mid-line incision, the first in January, 1978, and the second in October, 1978. No particular abnormality was noted in the abdominal cavity. The colon was palpated, and one polyp was found on the mesenteric side of the sigmoid colon, as noted in the barium enema in both cases. In the second case, an intra-operative sigmoidoscopic examination was performed, and the presence of a polyp was confirmed in the sigmoid colon 30cm from the anus. Polypectomy was performed through an incision of the antimesenteric side of the colon. Both patients were followed, and no further symptoms or new lesions were discovered. The excised polyps were oval, 1.5×1.5 cm in size in the first case and 1.0×1.5 cm in size in the second case. The surface was smooth, eroded and dark red, and the consistency was solid. Each polyp, had a short stalk and hemorrhage was noted on the surface.

Histopathological findings

Specimens from both polyps were first reported to be adenomatous polyps on histological examination. Although grossly they seemed to be adenomatous polyps, the microscopic appearance differed from that of typical adenomatous polyps usually found in adult colons. The histological appearance in these cases was characterized by proliferation of glandular

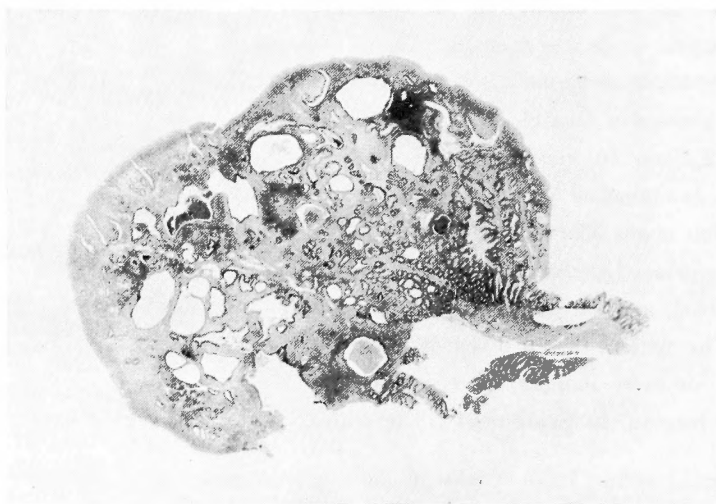


Fig. 2. Low power view of juvenile polyp. The stroma with hemorrhage is abundant, and dilated glands contain mucus and cellular debris. (Case 2)

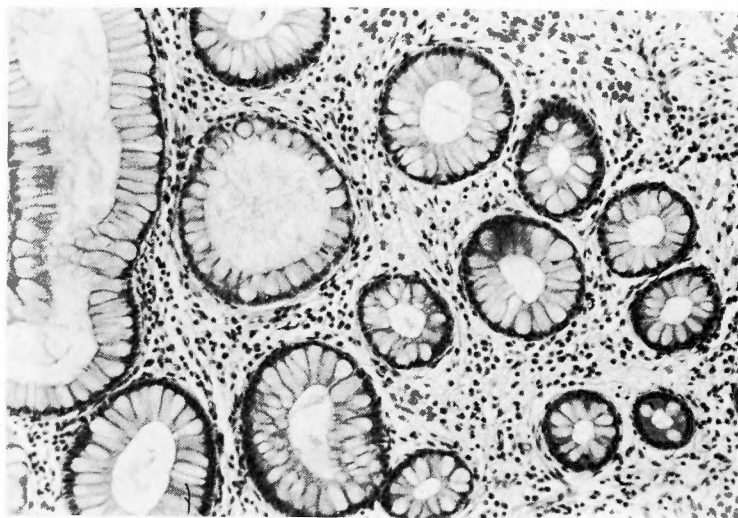


Fig. 3. High power view of juvenile polyp. Cystic spaces are lined by actively mucus-secreting uniform columnar cells. Stroma is infiltrated with inflammatory cells. (Case 2)

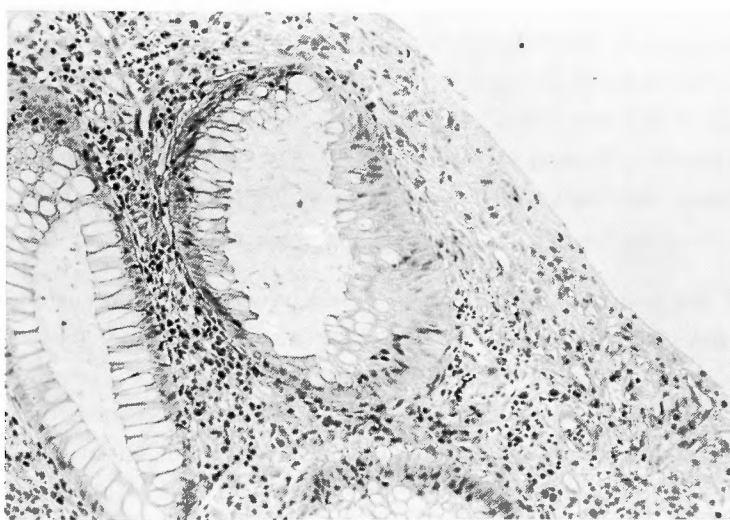


Fig. 4. Surface of juvenile polyp. Ulceration of the surface and many inflammatory cells in the stroma. (Case 1)

and stromal elements associated with marked vascularity, cellular infiltration with lymphocytes, plasma cells and polymorpho-nuclear leukocytes, with numerous eosinophiles and some focal area of necrosis and hemorrhage, as shown in Figures 2 and 3. The denuded epithelium on the surface had a characteristic appearance (Figure 4). The actively secreting mucous glands, consisting of uniform columnar epithelial cells with small nuclei, were well-oriented and showed no mitotic activity. Goblet cells were numerous. The glandular lumen contained mucous secretion and sometimes polymorpho-nuclear leukocytes, epithelial debris and red cells. Adenomatous polyps typically have the following histological appearance : the epithelial

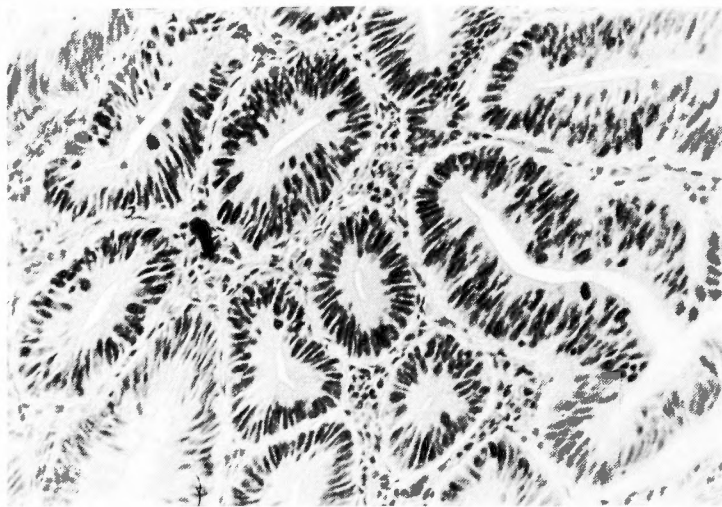


Fig. 5. High power view of adenomatous polyp in adult recognized in another case. The stroma is scant. The glands are hyperplastic but mostly regular in size. The nuclei are oriented basally and show relatively frequent mitoses.

elements resemble normal mucosa, except that the cells are more elongated and have a more darkly stained cytoplasm within which is situated a basally directed hyperchromatic nucleus. Secretory activity is less and mitotic activity is more frequent than in the normal mucosa, as shown in Figure 5. Because of this discrepancy in histological appearance, we agreed with the pathologist that these should be diagnosed juvenile polyps in both cases.

Discussion

Bleeding of the gastro-intestinal tract is a relatively common occurrence in children. SPENCER⁵⁾ analyzed over 900 records of one children's hospital and reported that in 476 cases there was gastro-intestinal hemorrhage: esophageal varices; esophageal, gastric or duodenal ulcer; diverticula of any portion of the gastro-intestinal tract, including Meckel's diverticulum; intussusception; malrotation; volvulus; gangrene, strangulation or infarction of any portion of the bowel; regional enteritis and ulcerative colitis; anal fissure; and other lesions of any portion of the alimentary tract associated with bleeding. The source of the hemorrhage in the 746 cases was identified as anal fissure in 83 cases, intussusception in 61 cases and polyps in 59 cases. Several authors have called attention to the fact that there are significant differences in the pathological and clinical behavior of polyps of the rectum and the colon between children and adults. Polyps of the colon and rectum in children were reported as adenoma by HELWIG et al.¹⁾ in 1946, and they mentioned the following points of difference between children and adults: relatively large amounts of stroma composed of connective tissue, frequently heavily infiltrated with inflammatory cells including plasma cells, lymphocytes and leukocytes, especially eosinophiles in children. Glandular structure with papillary epithelial growth and desquamated epithelium on the polyp surface were also characteristic histological features and their appearances described

were as the same in juvenile polyp, but at the time, juvenile polyp was not thought to be a clinical and pathological entity.

In 1960 KNOX et al.²⁾, established juvenile polyp clinically and pathologically as distinct from other polyps of the colon with rare potentiality of malignancy. In 1963 ROTH et al.³⁾, recognized juvenile polyp not only in children but also in adults and noted that the age distribution showed a biphasic pattern. The children's group included 62.6% of all cases, with an age range from 11 months to 10 years and an average age of 4.1 years, while the adult group had an age range from 17 to 61 years with an average age of 25.5 years. The common clinical features of juvenile polyp were rectal bleeding in 80%, protrusion of the polyp in 24% and abdominal pain in 6.3%. The most common location was the rectum (72%); 11% were in the sigmoid colon and 17% at other sites. The number of polyps ranged from one to approximately thirty; in 88% of the cases the polyp was solitary. The cases reported here met the clinical and pathological criteria, and we are convinced that they have the characteristic features of juvenile polyps. Although juvenile polyps appear to be cystic glands and have a large amount of stroma, it is possible that they are misdiagnosed as adenomatous polyps, since their architecture resembles adenomatous polyp histologically, if the entity of juvenile polyps is not known. At first, our cases were diagnosed as adenomatous polyp, then the diagnosis were corrected to juvenile polyp. It is supposed that the entity of juvenile polyp is not well-known to clinicians and pathologists in Japan and that the reason this disease is rarely recognized in Japan may be due not to the difference of race or geography but to the lack of knowledge of this disease in Japan. It is important to point out the existence of this entity and its clinical and pathological features. We must re-examine the records of rectal and colon polyps in children to determine the true incidence of juvenile polyps in Japan.

Summary

A 6-year-old and a 3-year-old boy had rectal bleeding without any other symptoms. Sigmoidoscopic and roentgen examinations showed solitary polyps in the sigmoid colon, and these were excised transabdominally. Histologically, these polyps were diagnosed as adenomatous polyps at first, but after careful observation, their histological appearance showing abundant stroma with infiltration of leukocytes, especially eosinophiles, and cystic glandular structure was considered different from that of adenomatous polyp, and the diagnosis was corrected to juvenile polyp. Juvenile polyps are not clinically and pathologically well-known in Japan, so it is possible that they are usually diagnosed adenomatous polyps.

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和文抄録

若 年 性 ポ リ ー プ の 2 例

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最近, 2例の若年性ポリープの症例を経験した。これらの2例は最初, 腺腫性ポリープと組織学的に診断された。しかし典型的な組織学的所見とは異っており, 表面の上皮の脱落, 盛んな粘液の分泌を伴った囊腫様の腺構造, 腺腔に比較して間質が豊富で, この間質には強い炎症細胞浸潤を伴い, 特に好酸球の浸潤が

特長的であった。これらの所見から, 腺腫性ポリープではなく, 若年性ポリープと判明した。この診断経過から, 日本で若年性ポリープの症例の報告が少ないのは, その実体が充分に知られていないため, 一見, 腺腫性ポリープに似た組織像を呈するために, 誤って診断されている可能性が強く示唆された。